The Ulster Medical Journal, Volume 58, No. 1, pp. 111–113, April 1989.

Case report

Papillary carcinoma arising in a thyroglossal cyst

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Accepted 5 January 1989.

Cysts of the thyroglossal duct are common lesions, the majority of which present as a midline swelling in the anterior aspect of the neck. Infection and sinus formation are not uncommon complications, but the development of malignancy within a cyst is very rare. To date only 116 cases have been documented in the world literature.[1, 2, 3] This report describes a papillary carcinoma arising within a cyst which presented clinically as a simple cyst.

CASE HISTORY

A 65-year-old man presented with a painless midline swelling in the front of the neck, which had been present for two weeks and had been increasing in size. Examination revealed a mobile 2·5 cm diameter cystic mass in the anterior midline of the neck in the hyoid region, which moved on swallowing. The thyroid gland was normal and no regional lymph nodes were palpable. Aspiration of the mass yielded 4 ml straw-coloured fluid and reduced the size of the lesion. Cytological examination revealed poorly cellular fluid, the cell population being composed largely of histiocytes. No epithelial cells were seen.

At surgery the cyst was exposed and mobilised down to the level of the hyoid bone. The central portion of the hyoid bone was removed and the cyst widely excised up to the floor of the mouth between the hyoglossus muscles.

The specimen was a 2·5 cm diameter cystic structure. Histological examination showed a thick focally hyalinised fibrous wall, with foci of thyroid follicles and scattered aggregates of lymphocytes. The cyst lining was thrown into papillae, composed of fibrovascular cores covered by cuboidal epithelium (Fig 1). In areas the epithelium showed multilayering, nuclear overlapping and optically clear nuclei. Many of the stromal cores contained calcospherites or psammoma bodies (Fig 2). The features were of a papillary carcinoma arising within a thyroglossal cyst. Epithelial invaginations were present in the fibrous tissue but there was no evidence of invasion beyond the cyst wall. There were no metastases in a small draining lymph node.

A post-operative isotope thyoid scan showed normal and symmetrical uptake in both lobes of the thyroid gland. There was no evidence of a lesion within the thyroid gland and no ectopic thyroid tissue was identified.

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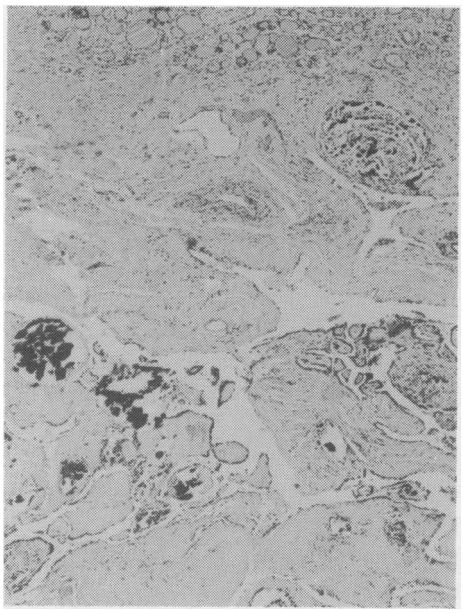

Fig 1. Thyroglossal cyst wall and papillary tumour in lumen (H&E, x 120)

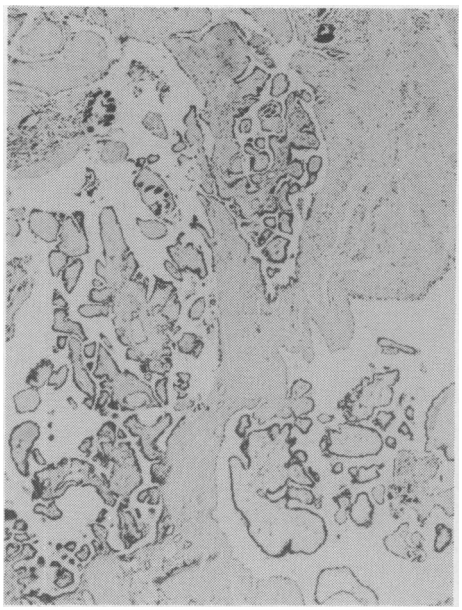

Fig 2. Papillary carcinoma with psammoma bodies (H&E, x 120)

DISCUSSION

Thyroglossal cysts are embryological abnormalities resulting from the persistence of the thyroglossal duct left by the descent of the thyroid gland. Cysts may form at any point along the line of descent from the base of the tongue to the retrosternal region, but most occur between the hyoid bone and the thyroid gland. The lining epithelium varies according to the site and may be squamous, cuboidal or columnar in type. The majority contain ectopic thyroid tissue and aggregates of lymphocytes deep to the epithelial lining.

While thyroglossal cysts are commonly complicated by infection and sinus formation, the development of malignancy is rare. An incidence of 1% is given by Allard.[4] In a review of the literature, 114 cases were identified[2] and more recently two further cases have been reported.[2,3] Malignancy may develop at any age but there is a predilection for females. Papillary carcinoma is the most usual histological variant, accounting for 83% of cases, and it has been suggested that the papillary carcinoma represents malignant metamorphosis in ectopic thyroid tissue.[2] Squamous cell carcinoma was found in seven of the 117 reported cases (6%). This arises in either a cyst high in the neck lined by squamous epithelium or in metaplastic squamous epithelium in an inflamed cyst situated lower in the neck. The remainder include adenocarcinoma, follicular carcinoma and mixed papillary-follicular carcinoma. Medullary carcinoma has never been reported in a thyroglossal cyst, perhaps because no parafollicular cells are found in ectopic thyroid tissue. Special stains were performed on this case and no parafollicular cells were identified. A pre-operative diagnosis of malignancy is rare and aspiration cytology proved unhelpful in this case.

The standard treatment for a benign thyroglossal cyst is the Sistrunk procedure. This remains adequate treatment if a papillary carcinoma is diagnosed on

histological examination and is found to be confined to the cyst, although some authors advocate further surgery if the tumour has breached the cyst wall.[4, 5] Metastatic spread is very uncommon. It is reported in 8% of cases,[4] with deposits occurring in the liver and lungs.

Joseph and Komorowski reviewed 52 cases of thyroglossal duct carcinoma, three of whom died as a result of extensive local recurrence or metastatic disease to the liver and lungs.[6] Two of these patients had a tissue diagnosis of papillary carcinoma and the third a diagnosis of squamous carcinoma. It is felt by some authors that the prognosis of papillary carcinoma in a thyroglossal cyst is similar to that of tumours arising in the thyroid gland.[1] Squamous carcinoma appears to have a considerably worse prognosis.

I am grateful to Mr M Bell and Mr V Loughlin for permission to report this case.

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